

UNILATERAL RIB-NOTCHING FROM THE COLLATERAL CIRCULATION AFTER DIVISION OF THE SUBCLAVIAN ARTERY

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The operation of subclavian-pulmonary anastomosis has provided another cause for unilateral rib-notching—the collateral circulation to the arm after division of its subclavian artery. The flow is, of course, in the reverse direction to the flow in the collateral arteries that by-pass a coarctation of the aorta.

We are reporting three cases in which there was striking unilateral rib-notching after a subclavian-pulmonary anastomosis, in one with other signs of a collateral circulation in the chest wall. My attention was drawn to the first of these patients by Dr. Curtis Bain as long ago as 1949 (Fig. 1A), but I did not then see the explanation. I thought there might be some new collateral circulation to the lungs, for small arteries from the mediastinum had often been seen entering the hila in patients with Fallot's tetralogy. Barrett and Daley (1949) also drew attention to the arteries they had seen at this site coming from the mediastinal, phrenic, and internal mammary arteries.

It was not until several years later when this boy was re-examined and two further patients were seen, all of whom had rib-notching on the side where the subclavian artery had been divided, that the correct explanation became apparent. The point at which the subclavian is divided is not far from the usual site of a coarctation, and anastomotic vessels that can carry blood from some branches of the subclavian and axillary arteries to parts below the coarctation can do so equally well in the opposite direction, from the lower part of the chest wall to the axillary artery and the arm.

When discussing this conclusion with my radiological colleagues, Dr. Hills and Dr. Dow, I found that unilateral rib-notching after division of a subclavian artery had been described by Kent (1953). He was, I think, the first to link these two features and to suggest the right explanation. He reported two cases, both with rib-notching on the side of the operation. The first had a complex lesion, probably dextrocardia and tricuspid atresia, and the second did not have an anastomosis completed: but, as he says, this does not affect the argument, because the subclavian artery was divided in both—for a successful anastomosis in the first and for an attempted anastomosis in the second. Since his paper this syndrome has been observed by several radiologists: it does not, however, seem to be so widely known among physicians.

As will be seen later, the condition is much more common than has been supposed, though generally in a less striking form than in these three cases. Sometimes there are also signs of the collateral circulation in the chest wall. Thus in Case 1, there was a prominent pulsating artery passing over the right clavicle very similar to the one shown by Kent (1953, his Fig. 10) as well as arteries in the chest wall over the lower ribs.

CASE NOTES

To save repetition, some features common to all three patients will be given first. All three had subclavian-pulmonary anastomosis done by Sir Russell Brock in 1948 and all have developed

striking unilateral rib-notching on the side on which the subclavian artery was divided. Two had Fallot's tetralogy and were severely disabled (Cases 1 and 3) and the third who had tricuspid atresia was moderately disabled (Case 2). All three had been deeply cyanotic from infancy and all were polycythæmic, with the hæmoglobin raised to between 140 and 150 per cent and with clubbed fingers and toes. None had any signs of coarctation of the aorta and the highest systemic blood pressure was 105/75 mm. All three squatted before, but not after operation.

All were greatly improved after the operation and have kept well for nine years, so that they can now lead normal quiet lives and earn their living, one as an architect, one as a warehouseman, and one as a laboratory assistant. All have a continuous murmur over the site of the anastomosis.

In none of them was there any visible rib-notching in the films taken before operation. It is, I think, no more than coincidence that they all obtained very good results from the operation and that two of them had very small hearts before it (Cases 1 and 2) for these are not uncommon with morbus cœruleus.

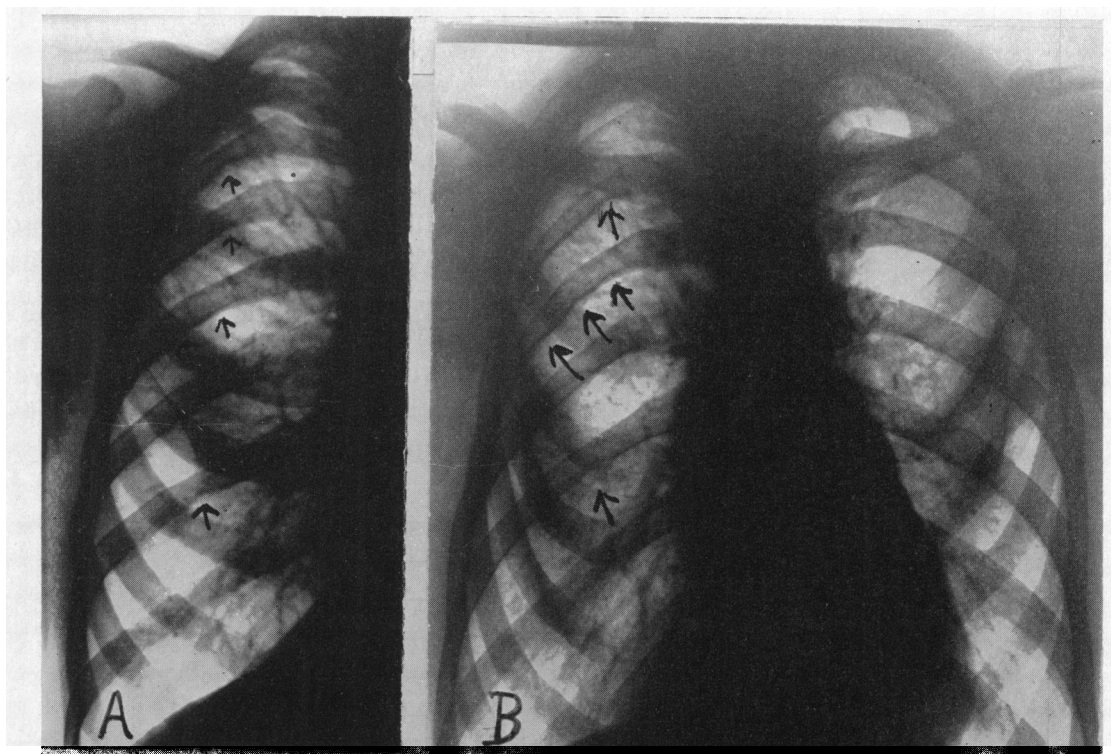


FIG. 1.—Case 1. Unilateral rib-notching on the right after a right-sided subclavian-pulmonary anastomosis for Fallot's tetralogy in 1948. (A) In 1950, (B) in 1957. Both show decisive rib-notching on the 4th, 5th, 6th, and 8th ribs. The heart is not enlarged (c.t.r. 48 per cent).

Case 1. A boy, aged 13, had Fallot's tetralogy and could rarely walk more than twenty yards. He had a right-sided anastomosis in June, 1948. His improvement came slowly and at first we were afraid there might have been some thrombosis in the anastomosis but within three months the result was excellent and he was walking a mile and cycling.

In October, 1949, Dr. Curtis Bain, who had been seeing the patient for years, found a striking collateral circulation that had not been visible before and seemed to involve the internal mammary, vertebral, and intercostal arteries on the right side, with some rib-notching on this side. When seen by us in 1950 he was leading a normal life at school, except for games. He was much less cyanosed and polycythæmic and his hæmoglobin percentage was nearly normal. Large pulsating collateral arteries could be seen and felt over the clavicle and sternum, but we were less certain about the rib-notching on the X-ray film (Fig. 1A). The

femoral pulse was easily felt, the blood pressure normal, and there was no evidence of coarctation or of a past thrombotic obstruction of his aorta. I did not, at that time, think of the correct explanation.

Nine years after the operation he is extremely well, working regularly in a laboratory and leading a normal quiet life. The cyanosis is slight and the hæmoglobin has fallen from 148 before operation to 114 per cent. The heart is larger but still within normal limits for it had been very small before (c.t.r. 41% in 1948, 48% in 1950 and in 1957). The superficial collateral circulation is less obvious as he has put on weight, but can easily be seen and felt over the lower rib spaces and where a prominent artery passes across the right clavicle. The rib-notching is now more obvious (Fig. 1B) mainly on the 4th, 5th, 6th, and 8th ribs on the right side (Case PO56).

Case 2. A man, aged 20, had tricuspid atresia but only moderate disability as he could walk a mile at his own pace. He had a left-sided anastomosis in 1948. After nine years he is working as a warehouseman and on a recent holiday walked twelve miles—one of the best results I have seen in tricuspid atresia. His small heart has not increased any more than usual (c.t.r. from 40 to 45% one month after operation and 44% nine years after). He still has some cyanosis and clubbing of the fingers and the hæmoglobin has not fallen greatly and is about 130 per cent.

In his case there is less collateral circulation visible over the chest but there is deep rib-notching of the 8th left rib and less obviously of some others (Fig. 2B). In retrospect, this could be seen in the X-ray of 1951 (Fig. 2A) but not in that of 1949, the year after his operation. We are indebted to Dr. W. Whitaker of Leeds for seeing him again and sending us the last X-ray film (Case O278).

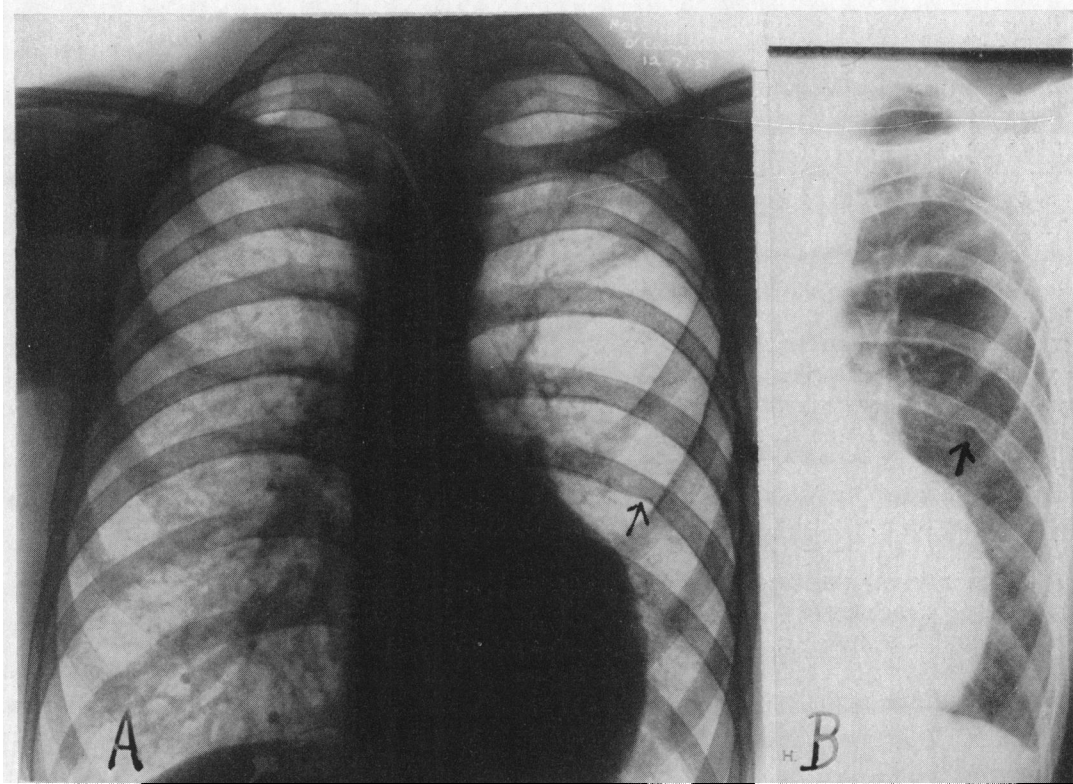


FIG. 2.—Case 2. Unilateral rib-notching, mainly of the 8th left rib after left-sided subclavian-pulmonary anastomosis for tricuspid atresia. (A) In 1951, (B) in 1957. The heart was small before, and remained so after operation, although this was most successful. The 7th left rib in (A) shows minor changes that we have found alone in many cases (see text).

Case 3. A boy, aged 12, had Fallot's tetralogy and could not walk more than 100 yards. After a left-sided anastomosis in 1948, he improved greatly. In 1951 he was attending school regularly and could walk a mile; he was less cyanosed, and the hæmoglobin had fallen from 148 to 104 per cent. The notching on the 5th left rib was not noted (Fig. 3A).

In 1957 he is still doing well and working as an architect. His heart is larger (c.t.r. 57% from 53% in

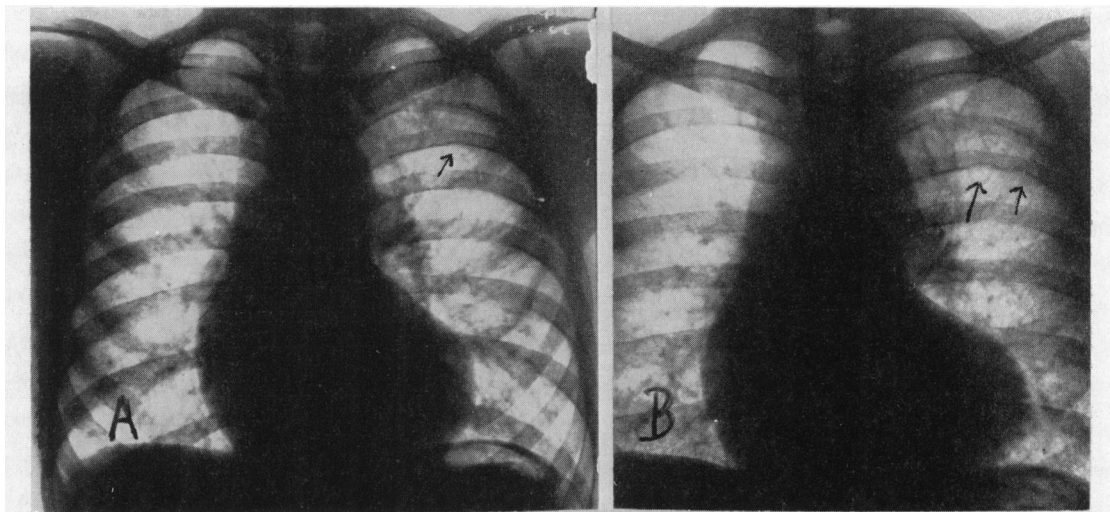


FIG. 3.—Case 3. The development of rib-notching. (A) Four years after a left subclavian-pulmonary anastomosis for Fallot's tetralogy the notches were not detected, but are visible in the 5th left rib below the rib that has regenerated. (B) Five years later the rib-notching is deeper. Incidentally the heart has become somewhat larger (c.t.r. 57% instead of 53%).

1951 and 48% in 1948) and perhaps this may have helped in the detection of the rib-notching at mass radiography but it was also deeper (Fig. 3B). We are indebted to Dr. Posner of Stoke-on-Trent for observing this and for sending us the X-ray film (Case O664).

THE ARM AFTER DIVISION OF THE SUBCLAVIAN ARTERY

A good collateral circulation develops in many parts of the body after loss of the main blood supply. This is so in the arm after division of the subclavian artery, and any significant trouble after a Blalock operation is rare. For a month or two the arm may be rather colder than usual and the patient may be disinclined to use it, but more serious troubles are rare though they have been reported. Holling (in Campbell, 1948) found that two weeks after the operation the forearm blood flow per unit volume was reduced, both at rest and with exercise, but after two months (and once after three weeks) there was no difference either at rest or with exercise. Generally the clubbing of the finger improves equally in both hands but occasionally more so on the side where the subclavian has been divided, suggesting that in these cases the flow may not have returned to a normal level (Campbell and Deuchar, 1953).

A few months after a Blalock operation, most patients are unaware of anything being different in the arm on the side of the operation, though even after eight or nine years I have seen two or three who find it a little weaker than the other, and rarely it is a little smaller. The radial and brachial pulses do not become nearly as strong as before, and up to four years after operation Campbell and Deuchar (1953) were unable to measure the blood pressure by auscultation in the arm on the side of the operation in any patient. As the years pass it is possible to measure the systolic pressure by palpation more often but very rarely the systolic and diastolic pressures by auscultation, though after nine years I have obtained readings that agreed with those in the other arm in one patient and were only 20 mm. below them in one other.

THE COLLATERAL CIRCULATION TO THE ARM

(a) *After Division of the Third Part of the Subclavian Artery.* A century ago when peripheral aneurysms were common, division of a subclavian or other large peripheral artery was practised more frequently. Astley Cooper, famous for his contributions to arterial surgery (Brock, 1952), was deeply interested in the collateral circulation. I wondered if these early accounts would include any reference to the intercostal arteries. The first number of the Guy's Hospital Reports

in 1836 contains papers on the circulation that had developed several years after Astley Cooper had ligated the external iliac and common carotid arteries for peripheral aneurysms. There is also an excellent account by Key (1836) of the collateral arteries that were found at necropsy in a man who had undergone an operation for an axillary aneurysm twelve years before.

In such operations the branches of the subclavian are available as a main "providing" source for the collateral circulation. Even so, Key's illustrations show that the collateral arteries extended a long way down the chest wall, certainly as far as the fifth rib. The main anastomosing arteries were (1) posteriorly, from the suprascapular and posterior scapular (from the thyro-cervical) to the infrascapular and so to the axillary artery; (2) internally, from the internal mammary to the short (superior) thoracic, long (lateral) thoracic, and infrascapular (subscapular) arteries, and so to the axillary artery; and (3) smaller but very tortuous branches in the axilla.

Atkins and Joseph (1955) have investigated the collateral circulation in a man who died nineteen years after the right subclavian had been ligated for hæmorrhage; their findings, which were confirmed by radiograms of the injected collaterals, were very similar.

(b) *After Division of the First Part of the Subclavian and its Branches.* The position after a Blalock operation is different, for the subclavian is divided nearer to its origin and the branches from the first part—the vertebral, the internal mammary, the thyro-cervical, and generally the costo-cervical arteries—have to be ligated and divided to provide an adequate length of subclavian for the anastomosis, since the free portion of the first part is generally no more than one inch (2.5 cm.) in length and often only half this, even in an adult. The branches cannot, therefore, help directly with the collateral circulation, though they may do so indirectly if they develop fresh anastomotic connections at their proximal ends beyond the division.

Sir Russell Brock has generally found division of all these branches necessary and at operations carried out in 1948 this was almost certainly the case. He has kindly looked up his operation notes and finds that in Case 1 all these branches were divided, in Case 2 the vertebral artery certainly and probably the other branches, and in Case 3 the subclavian was divided just beyond the vertebral and internal mammary which were almost certainly ligated.

When, as is usual, none of these branches is available, the collateral circulation can arise only from (a) branches of the aorta, (b) branches of the carotid, and (c) arteries from the other side of the body. One important route is from the aortic intercostals to the anterior intercostals and so to the internal mammary, and by its anastomoses in the chest wall to the superior thoracic, lateral thoracic, and subscapular branches of the axillary artery. This would, of course, explain the presence of large tortuous intercostal arteries and the consequent rib-notching in one side. I regret that I have missed several opportunities of examining the collateral circulation after injection at necropsy and this should be done.

Webb and Burford (1952), reporting one of the rare cases where the arm had become gangrenous after an operation discussed also anastomoses from the superior thyroid, occipital, and muscular branches of the external carotid to the inferior thyroid, suprascapular, and transverse cervical arteries, and so through anastomoses round the shoulder to the subscapular and other branches of the axillary artery. They give a useful diagram of the possible sources of the collateral circulation. Incidentally, they stated that in this case all the branches of the subclavian had been divided though generally they had not found this necessary.

(c) *From the Arm in Coarctation of the Aorta.* Bramwell and Jones (1941) gave an excellent account of the collateral circulation in coarctation. If the vertebral, internal mammary, thyro-cervical, and costo-cervical arteries have been rendered useless for the collateral circulation, the only remaining channels in their list available to function in a reverse direction would be between the lateral and dorsal branches of the aortic intercostals on the one hand and the subscapular, superior thoracic, and lateral thoracic branches of the axillary artery on the other hand.

THE FREQUENCY OF UNILATERAL RIB-NOTCHING

These considerations about the collateral circulation explain why the intercostal arteries may become tortuous enough to produce notching of the ribs. It seemed, in fact, rather surprising that

it was not much more common after subclavian-pulmonary anastomosis. Until recently I thought it could not be common, for hundreds of films that had been examined by myself and many others, including radiologists, had revealed only these three cases.

When most of this paper was written, I examined with special care, 16 consecutive cases that had a subclavian-pulmonary anastomosis more than eight years before. There was no case with unequivocal rib-notching that had been missed, but many showed some slight irregularity of the lower border of one or more ribs, always on one side only, that might have been due to a large intercostal artery. Having recognized this possibility, I re-examined the same films a day or two later and now marked 9 of the 16 as showing slight unilateral rib-notching.

Dr. Dow kindly examined these independently and was even more certain that several of them showed unilateral rib-notching: he rejected one of mine but added one I had rejected, so that evidence of it was seen by both of us in 8 and by each of us in one other, making 10 altogether. In all these 10 the side on which we had seen the irregularity of the lower rib margin was found to be the side on which the subclavian had been divided. This convinced us that what we had seen was due to one or more enlarged tortuous intercostal arteries confined to one side, and that a minor degree of unilateral rib-notching is quite common after a subclavian-pulmonary anastomosis. The changes are so small that they are difficult to show in illustrations but perhaps the 7th left rib in Fig. 2A, the rib above the one with obvious rib-notching, gives an idea of the roughened lower rib margin that we are discussing.

UNILATERAL RIB-NOTCHING IN GENERAL

(a) *In Coarctation.* This condition is well-known and occurs when the left subclavian artery is involved in the coarctation or arises below instead of above it. The blood pressure in the left subclavian is then low, so that there would be less advantage in an anastomosis between it and the lower part of the body, and the anastomosis develops on the right side only. King (1937) reviewed fifteen such cases and added some of his own, but the case of Bayley and Holoubek (1940) was apparently the first in which unilateral rib-notching was seen. Campbell and Baylis (1956) reported two examples among their 130 cases of coarctation and one was illustrated in their Fig. 6A. MacLaughlin (1951), reporting another case where there was also notching of one scapula, discussed the causes of unilateral rib-notching without including any example of the type described by Kent (1953) and in this paper.

(b) *In Fallot's Tetralogy and Morbus Cæruleus.* Rib-notching, generally of slight or moderate extent, has been reported in cases that have had no operation, and often this has been mainly or completely unilateral. Batchelder and Williams (1948) described a case with notching of the 5th to 8th ribs on the left side, associated with Fallot's tetralogy and cystic disease in the left upper lobe: there was no coarctation at necropsy and it was thought to be due to an intercostal and mediastinal anastomosis.

In our series of cases of morbus cæruleus, four have been indexed as having unilateral rib-notching. In the first, a boy aged 10 with Fallot's tetralogy, the notching was deepest on the 5th and 6th right ribs but also visible on the 4th right rib (Fig. 4). Dr. Hills' report on the angiocardio-gram was "The arterial supply to the right upper lobe is abnormal and blood appears to reach this region from branches of the right subclavian artery". Certainly these branches filled about the same time as the subclavian and probably came from a superior intercostal that had more anastomoses than the usual ones with the first and second aortic intercostal arteries. Even in the patient just quoted without Fallot's tetralogy, Atkins and Joseph (1955) found two branches from the subclavian passing into the lung tissue itself. Our patient had a successful subclavian-pulmonary anastomosis on the left side and no evidence of coarctation was found.

In the second, a man, aged 30, with Fallot's tetralogy, large tortuous intercostal arteries were seen in the upper left interspaces below the rib-notching. At the age of 10 fluid had been removed from his left pleural cavity and he maintained that he had been less cyanosed and less disabled since then. Part of his angiocardio-gram at 4 seconds was reproduced by Lowe (1953, Fig. 6).

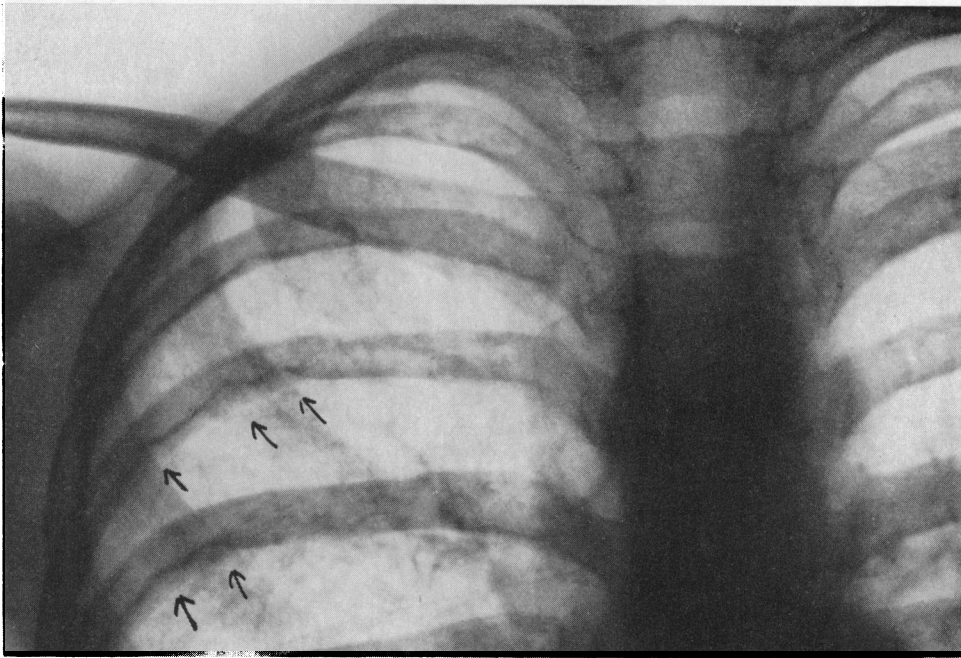


FIG. 4.—Unilateral rib-notching on the 4th rib and deeper on the 5th and 6th right ribs, in a boy, aged 10, with Fallot's tetralogy, before operation. Angiocardiography, showed unusual filling of the right upper part of the lung, which seemed to come partly from the subclavian artery.

In the third, a woman, aged 35, who was thought to have Ebstein's disease, there was notching of three of the upper right ribs (Fig. 1A, Reynolds, 1950). In the fourth, a girl, aged 16, the diagnosis was pulmonary atresia (or possibly truncus arteriosus) and there was a right-sided aortic arch and a continuous murmur that was maximal on the right side but widely conducted. There was slight notching of the fifth and deep notching of the sixth right ribs (Fig. 5) and perhaps of the seventh and tenth. No other collateral circulation was found, nor any evidence of coarctation.

(c) *In other Conditions.* McCord and Bavendam (1952) reported one case in which the rib-notching was due to intercostal veins that had become dilated and tortuous from long-standing obstruction of the superior vena cava by a mediastinal mass. Although this is generally regarded as very rare, Dr. Dow tells me that he has seen two such cases, associated with thrombosis of the subclavian or superior vena cava: the cause of the thrombosis was tuberculous glands in one and was unknown in the other.

SUMMARY AND CONCLUSIONS

We have reported three striking examples of unilateral rib-notching after subclavian-pulmonary anastomosis, two for Fallot's tetralogy and one for tricuspid atresia, all on the side where the subclavian artery has been divided. Though this obvious notching is not common we have given reasons for thinking that a minor degree of it from large intercostal arteries is quite common.

This unilateral rib-notching is due to the collateral circulation to the arm of which the subclavian has been divided. When the first part of the subclavian is divided the branches are not fully available for the collateral circulation. Under these conditions, an important source of the blood supply is from the aortic intercostal arteries to the anterior intercostals and so to the trunk of the internal mammary, and by its anastomoses in the chest wall to the superior thoracic and lateral thoracic branches of the axillary artery.

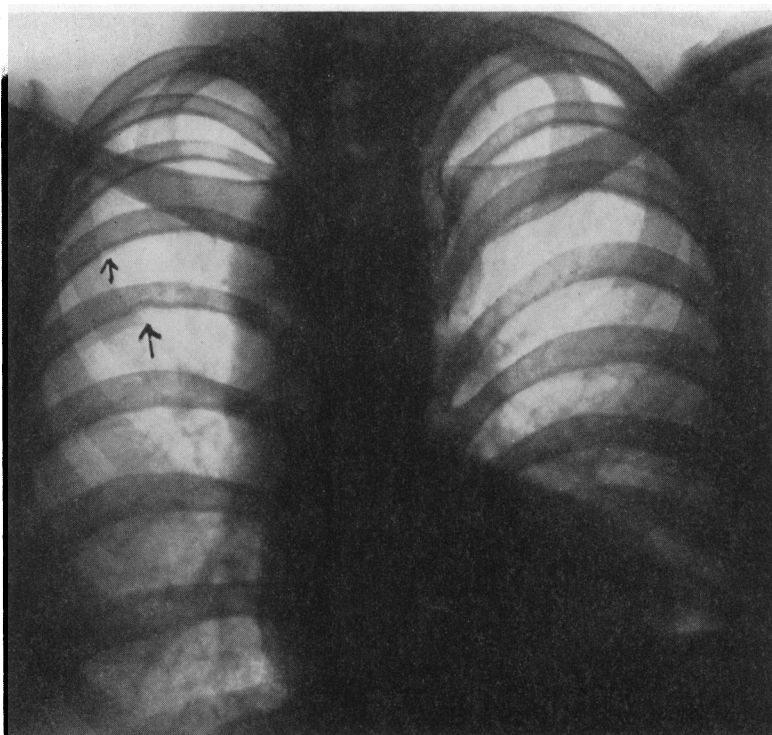


FIG. 5.—Unilateral rib-notching, shallow in the 5th and deep in the 6th right rib. From a girl, aged 16, with pulmonary atresia, who had no evidence of coarctation.

The collateral circulation that develops after subclavian-pulmonary anastomosis is compared with that after division of the third part of the subclavian, an operation that used to be done more frequently a century ago when peripheral aneurysms were common.

Other causes for unilateral rib-notching are discussed shortly.

A fourth striking example with unilateral notching of the 3rd and 4th left ribs has been seen since this was written.

I would like to thank Dr. J. D. Dow for his help and also for examining these radiograms, and Sir Russell Brock, whose surgical treatment has made these observations possible. I am indebted to Dr. Curtis Bain who drew my attention to the first of these cases, to Dr. W. Whitaker and Dr. E. Posner who let me have recent films of Cases 2 and 3, and to Mr. Engel of the Photographic Dept., Guy's Hospital, for the photographs.

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